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CASE REPORT

Multiple teeth showing invasive cervical resorption – an entity with little known histologic features

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Invasive cervical resorption is a relatively uncommon form of external root resorption, characterized by resorption of the cervical region of the root. There is progressive loss of cementum and dentine with replacement by fibrovascular tissue derived from the periodontal ligament, with deposition of cementum-like hard tissue. In most cases, a single tooth is involved. We report a case of invasive cervical resorption that resulted in loss of multiple teeth from two different quadrants. The case highlights the diagnostic difficulty that may arise in this uncommon lesion, the pathologic features of which may be mistaken for a fibro-osseous lesion or a low-grade sarcoma.

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A 33-year-old female was noted to have 'alarming erosion' of the root mass of the upper left incisors and canine on a routine radiograph. The patient had a repair of an incomplete cleft palate with an autogenous bone graft as an infant, and again at 17 years. A periapical radiograph of the area demonstrated extensive resorption of the roots of the upper left incisors and canine, which were clinically vital (Fig. 1a). The involved teeth were removed and the underlying bone curetted. Over the next 3 years, there was radiographic evidence of extension of the aggressive resorption process to the upper left premolar region, the upper right molar region, and the upper right premolar region, resulting in loss of eight teeth in total.

The tissue removed initially was fragmented and it was only at subsequent resections that the gross relationship of the soft tissue to the teeth became clear. The crowns of the extracted teeth were separated from the roots by soft fibrous tissue that had almost amputated the crown (Fig. 1b). At low power, the presence of tunnels of fibroblastic tissue burrowing into the dentine could be appreciated (Fig. 2a). The histologic features of the soft tissue revealed a variety of patterns. There were areas of fibroblastic proliferation lacking inflammation, some of which were quite cellular but without pleomorphism or mitotic activity (Fig. 2b). Some areas were more myxoid and others more fibrous in appearance. The more fibrous tissue contained focal cementicle-like structures (Fig. 2b) merging with occasional large irregular areas resembling woven bone (Fig. 2c). In later resections, the tissue was less fragmented and it became apparent that there was a zoning effect with the most active cellular fibrous tissue present close to the thin rim of still preserved dentine and predentine layer, i.e. at the advancing edge of the resorptive process (Fig. 2d). The pulp was not inflamed. More peripherally (Fig. 2e), the tissue was less cellular and focally contained remnants of odontogenic rests. Peripheral to these, the fibrous tissue contained irregular hard tissue. Some of this was dense acellular hard tissue-like bone or cementum while other areas were more cellular with prominent osteoblastic activity.

Comments

Invasive cervical resorption affecting multiple teeth is a very rare entity and the fact that the process was not confined to a single tooth probably contributed to lack of clinical recognition of the lesion. Heithersay (1, 2) documented the histology of this condition noting the evolution from the early fibroblastic lesion progressing to the later fibro-osseous type lesions with extensive bone/cementum deposition in the resorbing tissues. The latter areas may be histologically indistinguishable from periapical/osseous dysplasia. Heithersay (1) also illustrates burrowing of channels, deep into the dentine that may connect more apically with the periodontal ligament. The resorption characteristically stops short of the pulp leaving a narrow strip of dentine and predentine. There may be occasional massive involvement of the pulp by fibro-osseous deposition.

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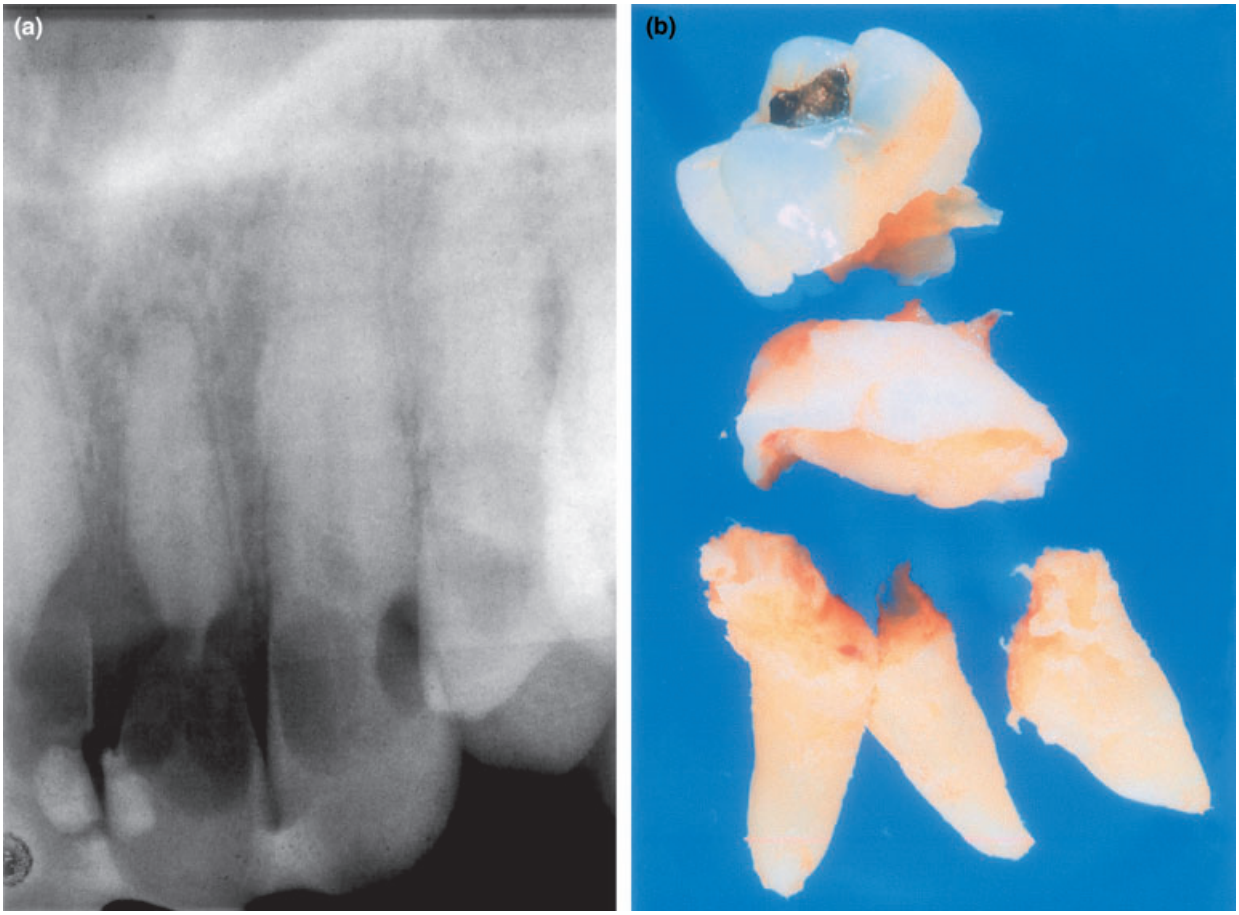

Figure 1 (a) Periapical radiograph at first presentation showing cervical resorption of incisors and canine; (b) crown and roots have become separated by soft tissue encircling the junction.

This case illustrates many of these features but highlights the difficulty in arriving at a diagnosis when the tissue is fragmented and the relationship of the various components to one another is lost. The appearances of the tissue when fragmented may suggest conditions such as fibrous dysplasia, cemento-ossifying fibroma or sclerosing osteitis. In 1960, Wade (3) described this condition as fibrous dysplasia of teeth. The presence of odontogenic rests might raise the possibility of an odontogenic fibroma or myxoma. The areas of very cellular stroma with bone formation might even suggest a low-grade osteosarcoma. Appreciation of the zonation of the different patterns is an important consideration in making the diagnosis, as is the case in conditions such as myositis ossificans. The initial tissue from this case was referred for several opinions to pathologists with bone/soft tissue and oral and maxillofacial expertise but all were baffled. The later material and evolving clinical picture prompted the correct diagnosis.

The presence of epithelium as seen in this case has not been emphasized previously. This most probably represents rest of Malassez because it is located in the region of the periodontal ligament.

The etiology of invasive cervical resorption is unknown but damage to or deficiency of the normally protective cementum layer appears to be necessary for initiation of the process (4). Despite the name, the resorptive process occurs at or below the cervical margin (5). A study of potential predisposing factors identified the following factors (in decreasing frequency): orthodontic treatment, trauma, dentoalveolar surgery, and intra-oral bleaching. Some patients had multiple factors, while 15% of patients in this study had 'no' predisposing factors (6). It is not clear why the resorptive process begins years after the initiating insult, although it has been suggested that a change in composition of the cementum make it less resistant to resorption that is induced later by a minor injury (7). It is possible that the resorption of multiple teeth in this case is related to the repair of the patient's cleft palate, a theory that gains support from the fact that only maxillary teeth have been affected. Pre-surgical orthodontic treatment, surgical flap procedures and placement of a bone graft could either independently or collectively have contributed to the condition in this patient.

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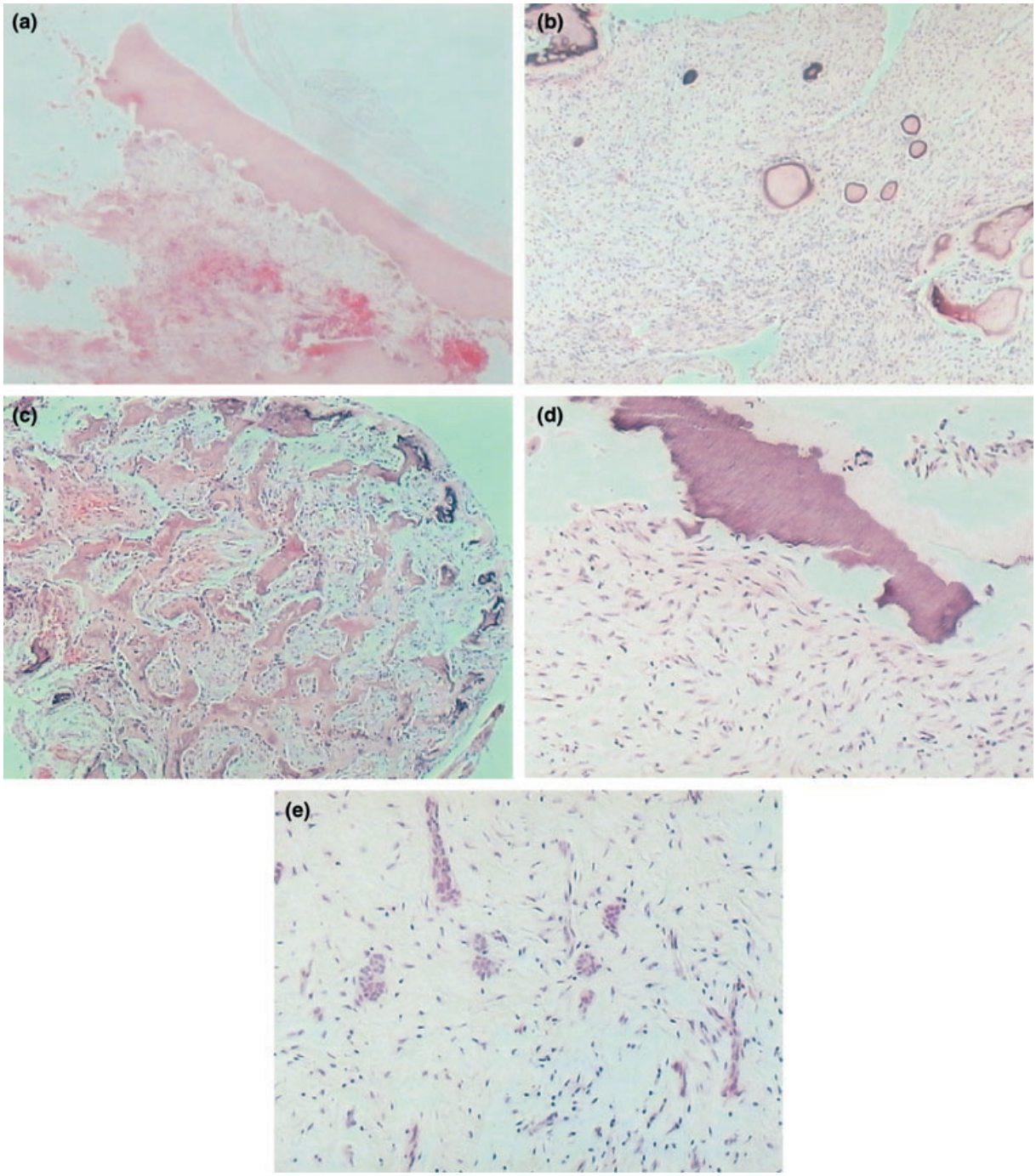

Figure 2 (a) Low power view showing burrowing effect of fibrous tissue with dentine either side; (b) uninflamed cellular fibrous tissue containing cementicles; (c) most peripheral zone with irregular deposition of variably calcified bone-like tissue resembling a fibro-osseous lesion; (d) fibrous tissue has eroded deeply into dentine; preserved pre-dentine layer is visible; (e) epithelial rests proliferating within fibrous tissue.

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